Intermittent claudication caused by a giant atypical lipoma of the thigh

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The main cause of intermittent claudication in lower limbs is peripheral vascular disease. Less commonly, the etiology can be extrinsic to vascular structures, as in the cases of tumors that, due to their rapid growth, can reduce the blood supply and produce intermittent claudication during gait. We report the case of a 49-year-old patient with intermittent claudication in the left lower limb, reporting the presence of a tumor in the inner side of the left thigh with rapid growth. Doppler and angiography magnetic resonance imaging examinations demonstrated the presence of an adipose tumor that was producing deep and superficial extrinsic compression of the femoral arteries. (J Vasc Surg 2012;56:808-11.)

Lipomas are benign encapsulated soft tissue tumors of mesenchymal origin that can appear at various sites. They represent approximately 10% of benign tumors; most of them are small in size and of low weight, and giant lipomas are uncommon.[1] Intermittent claudication has been estimated to affect around 15% of patients with chronic femoropopliteal disease aged over 60 years.[2] Atherosclerosis is the main etiology of peripheral arterial disease and an intrinsic cause of reduced arterial flow to lower limbs.[3] Intermittent claudication is rare in young patients without cardiovascular risk factors. If only one limb is affected, the main cause is extrinsic compression of arterial structures.[2] There have been rare reports of the alteration of arterial flow from causes extrinsic to the vessel, such as arterial compression by a tumor in surrounding tissues or popliteal entrapment.[4,5] Lipomas have been known to cause symptomatic obstruction of vascular structures, generally venous and occasionally abdominal viscera with a low resistance to compression.[6-9] We present the case of a 49-year-old patient with intermittent claudication in the left lower limb secondary to a giant subfascial lipoma localized between the deep and superficial femoral arteries.

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CASE REPORT

A 49-year-old woman was referred to our unit for a 3-month history of a large tumor in the medial left thigh. She had a history of obesity, hypertension, and had undergone breast cancer surgery 3 years earlier. The two cardiovascular risk factors were obesity (body mass index = 40.79 kg/m^2) and hypertension. The patient reported that pain in the left calf forced her to sit after walking for 250 meters. The pain was alleviated when she sat and bent from the hip. Physical examination demonstrated a smooth, well-delineated tumor of approximately 12 × 8 cm with elastic consistency in the femoral triangle (of Scarpa); it was adhered to deep planes and painless. Popliteal and pedal pulses of the lower limb were preserved and symmetric. There were no signs of venous insufficiency or hypertension. No deficit was found in the neurologic examination. At rest, the ankle-brachial index (ABI) was 1 in both lower limbs. After a treadmill test performed at 10° angle and at constant speed until claudication, the ABI remained at 1 in the right leg but fell to 0.46 in the left. Plain radiograph showed no significant alteration at bone level or presence of calcification within the soft tissue tumor. Magnetic resonance imaging (MRI) scans (Fig 1, *A* and *B*) revealed a deep subfascial adipose tumor between the sartorius, long adductor, and vastus intermedius muscles of the quadriceps. Contrast MRI indicated a tumor with heterogeneous areas that was closely related to the deep and superficial femoral arteries, whose diameters were reduced. Duplex scan examinations demonstrated peak systolic velocities of 89 cm/s in the proximal segment of the superficial femoral artery and 226 cm/s in the area of the superficial femoral artery subjected to maximum compression by the tumor, a ratio of 2.55; the peak systolic velocity was 79 cm/s at the popliteal artery. Our velocity criterion for the diagnosis of a significant arterial lesion was an increase in the acceleration in the segment that was at least 2.5 to threefold higher than the baseline acceleration in the same segment. No spontaneous venous flow was observed in the femoral duplex scan, but venous flow was observed after calf compression, ruling out deep vein thrombosis.

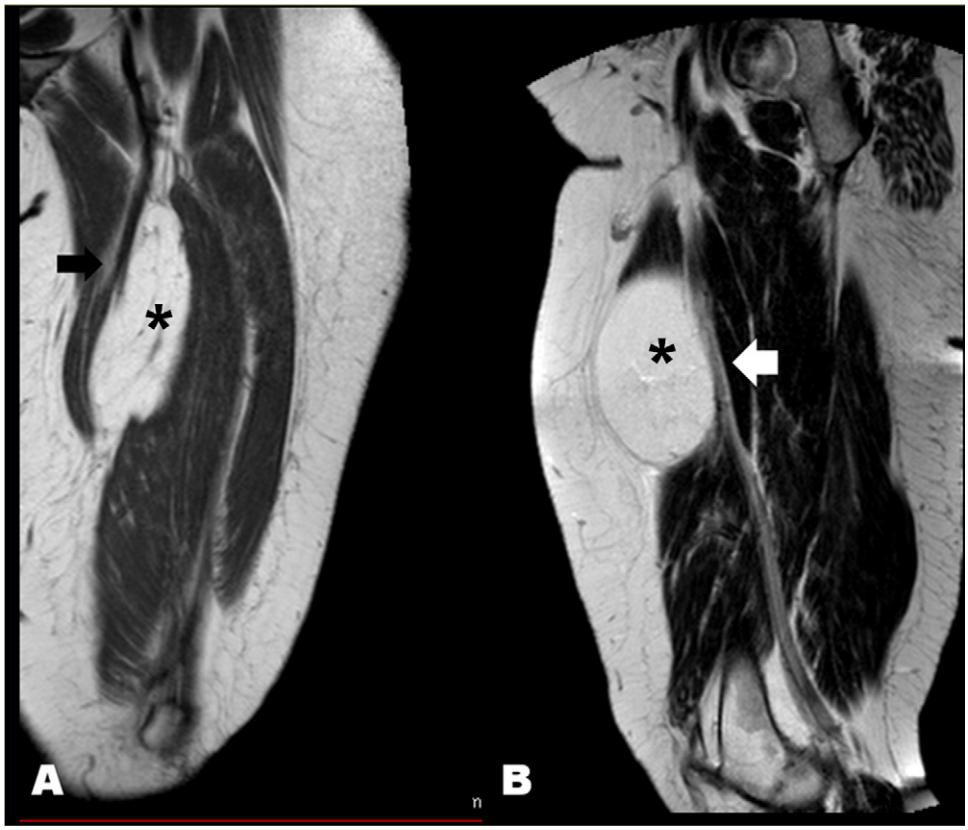

Fig 1. A, Coronal slice in T1, showing the relationship of the lipoma (*) with the femoral artery (*black arrow*) and thigh muscles. **B,** Sagittal slice in T2, showing the relationship between the adipose tumor (*) and femoral artery, which shows a slight narrowing of the vessel lumen (*white arrow*).

The patient underwent surgery to remove the tumor. Exposure of the tumor in deep fascia of the quadriceps revealed a nonlobulated encapsulated lipomatous mass of yellowish-brownish-gray color that measured 11 × 9 × 4.5 cm and was in intimate relationship with the femoral artery, vein, and nerve (Fig 2, *A*). Careful dissection of the internal compartment of the thigh revealed the deep and superficial femoral arteries, whose walls were adhered to the tumor capsule (Fig 2, *B*) but were not infiltrated by the tumor. After surgery, the patient recovered normal gait, and the intermittent claudication disappeared. Baseline and post-effort ABI at 1 month after surgery were compatible with normality. The histologic study confirmed that the mass was a benign lipoma (Fig 3).

DISCUSSION

A lipoma is a benign circumscribed tumor composed of well-differentiated adipocytes and is the most common soft tissue mass. Lipomas occur in 2% of the population and represent 50% of all musculoskeletal soft tissue tumors.[9] The tumor can originate at any site in the body that contains adipose tissue, but it typically arises in subcutaneous tissues of the upper half of the body and proximal limbs.[10] Superficial lipomas are more common than deep lipomas.[9] Intermuscular lipomas are rare (incidence of 1.8%) and have a recurrence rate of only 1% after resection, in comparison to 19% for intramuscular lipomas.[11] Tumors in the popliteal or inguinal region may compromise neurovascular bundles through encasement.[9] In the present case, a deep and unusually large intermuscular lipoma was located between the sartorius, vastus intermedius, and long adductor muscles of the medial left thigh with double extrinsic arterial compression, which produced intermittent claudication. In this case, the subfascial tumor produced a dynamic extrinsic compression of both arterial structures through the contraction of muscles and muscle fascia during gait. Except for esthetic motives, the only reason for removing a benign lipoma is to avoid complications derived from its compression of neurovascular structures, which is more frequent in subfascial tumors due to the limited distension capacity of the fascia.[6,7,10,12]

The prognosis of fast-growing subfascial lipomatous soft tissue tumors largely depends on their histologic type, ranging from nonproblematic benign lipomas to liposarcomas with regional and distant spread, which carry a poor prognosis.[13] When lipomas are larger than 10.0 × 5.0 × 3.0 cm, the differential diagnosis should include liposarcomas, especially if fast-growing. In MRI studies, the appearance of liposarcomas and lipomas is similar, although signs that characterize liposarcomas can occasionally be ob-

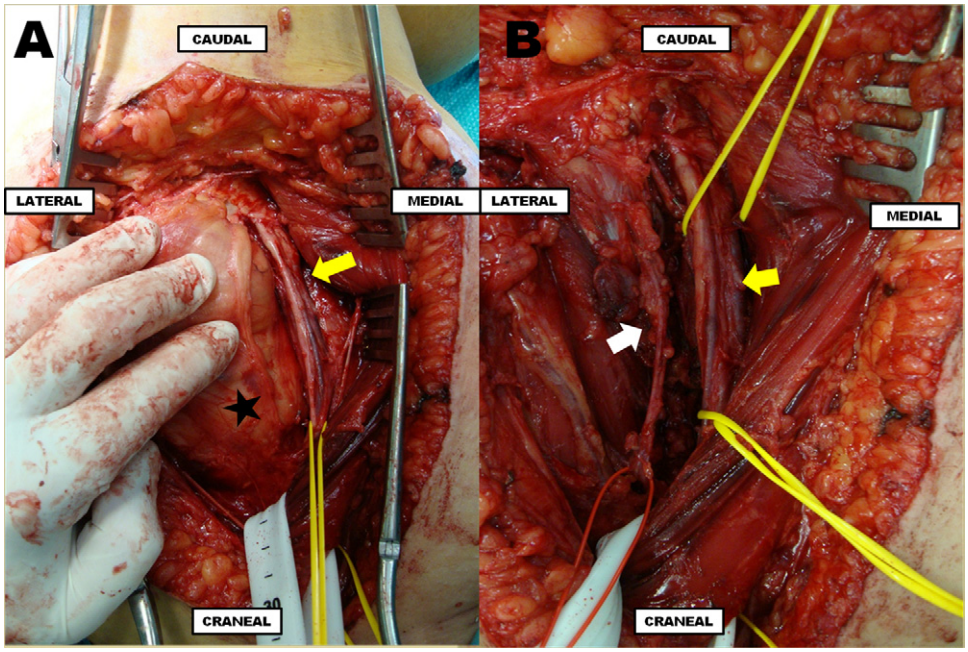

Fig 2. A, Image of surgical procedure: tumor in subfascial localization (*star*) and in close relationship with femoral vessels (*yellow arrow*). **B,** Image of surgical procedure: deep femoral artery (*white arrow*) and superficial artery (*yellow arrow*) after tumor resection.

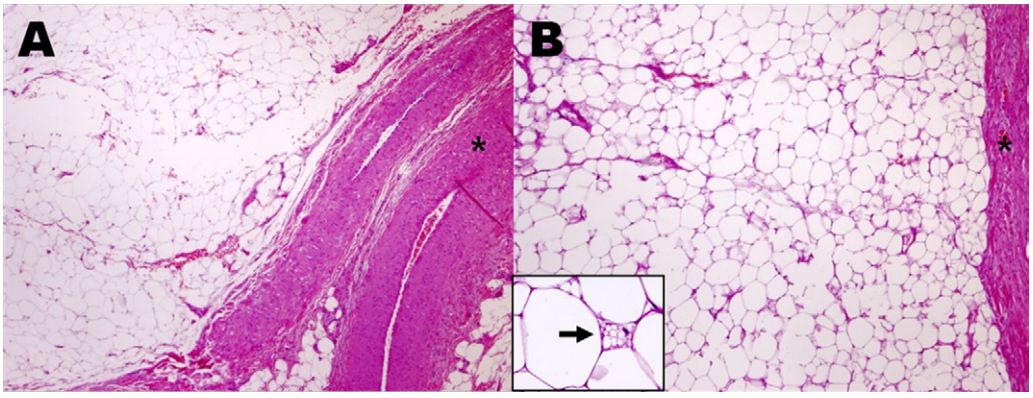

Fig 3. Histologic study. We highlight the presence of microvacuolated adipocytes (around 1%) and the absence of cell atypia or cell multinucleation. No mitosis or myxoid tissue was observed in the samples. **A,** Mature adipose tissue with microvascular network. Medium-caliber vessel showing compression and lumen narrowing (*) (hematoxylin-eosin [H&E] stain). **B,** Encapsulated univacuolated adipocytes of mature appearance (*) (H&E ×4). *Arrow*: multivacuolated adipocytes (H&E ×40).

served. In MRI studies, well-differentiated liposarcomas have thickened septae (>2 mm in thickness or with focal thickening) and nodular and/or globular areas of nonadipose tissue within the lesion or associated with the mass.[14,15] No such MRI signs suggestive of liposarcoma were observed in the present case.

The histologic study found no atypical cells or mitosis, ruling out the diagnosis of a liposarcoma. However, the observation of multivacuolated cells required a differential diagnosis to be performed with lipoblastoma, a benign tumor that appears between the ages of 3 months and 16 years. Histologically, this tumor is characterized by the presence of microvacuoles in mature adipocytes with scarce mitosis, gigantocellular cells, and myxoid tissue.[16] In the present case, the histologic and immunohistochemical study confirmed it to be a benign lipoma.

A computerized search of the literature from 1966 to December 2011, using MEDLINE, found no reports of intermittent claudication in a single limb produced by the extrinsic compression of a giant lipoma on both femoral

arteries at the thigh level. Hence, extrinsic vascular compression by a tumor is a very rare observation. Most reported cases of extrinsic compression are in veins, which have a thinner muscle layer in comparison to arteries and are consequently less resistant to vascular collapse. Extrinsic arterial compression in orthopedic surgery almost always has an iatrogenic etiology, although there have been rare cases of arterial compression in limbs produced by bone and soft tissue tumors.[1,4,17-19]

In conclusion, a hemodynamic study and complementary imaging tests (angiography MRI and duplex scan) are recommended in patients with intermittent claudication but no signs of intrinsic peripheral vascular disease in order to detect any extrinsic arterial compression produced by a tumor that may require surgical removal.

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